



CLINICAL REPORT

Escherichia coli osteomyelitis of the ischium in an adult

Amira Hamzaoui^{a,*}, Randa Salem^b, Mustapha Koubaa^c, Makram Zrig^c,
 Hichem Mnif^c, Abderazek Abid^c, Mondher Golli^b, Silvia Mahjoub^a

^a Internal Medecine Department, Fattouma Bourguiba Hospital, Monastir, Tunisia

^b Radiology Department, Fattouma Bourguiba Hospital, Monastir, Tunisia

^c Orthopaedic Department, Fattouma Bourguiba Hospital, Monastir, Tunisia

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KEYWORDS

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Summary Osteomyelites, bone infections of a hematogenous origin, are rare in the pelvis (2.3%) and are extremely rare in the ischium. Ischiatic osteomyelitis is usually found in children and adolescents, but has rarely been described in adults. The clinical presentation varies and the diagnosis is based on magnetic resonance imaging (MRI). The most frequently isolated germ is the staphylococcus, while *Escherichia coli* has been found in a few cases. We report a case of osteomyelitis from *E. coli* in a 46-year-old woman revealed by persistent fever. The point of entry was a septicemia from gastrointestinal origin, related to colon polyps. The clinical picture was also complicated by an antiphospholipid antibody syndrome (superior mesenteric vein and splenomesenteric branch thrombosis). The course was favorable thanks to appropriate antibiotic treatment and surgical debridement of the infection.

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Introduction

The ischium is a rare site for osteomyelitis [1]; it is generally diagnosed late at the stage of complications [2], and it is usually described in children and adolescents, but rarely in adults [3]. The clinical picture varies and diagnosis is based on nuclear magnetic resonance imaging (NMRI).

The most frequently isolated germ is staphylococcus, while *Escherichia coli* (EC) has been found in a few cases.

We report the case of EC ischiatic osteomyelitis with a gastrointestinal point of entry in a 46-year-old woman. The case is original because this diagnosis is rare, the presentation was limited to persistent fever with digestive disturbances, and biological results showed an inflammatory syndrome associated with an antiphospholipid antibody (APL) syndrome, resulting in abdominal deep venous thrombosis identified on CT scan. The ischiatic symptoms did not appear until 15 days later despite the administration of empiric antibiotics for pain in the left hip. Stage III ischiatic osteomyelitis was diagnosed on CT scan and NMRI. A few

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* Corresponding author. Tel.: +216 98616195.

E-mail address: hamzaoui.amira@yahoo.fr (A. Hamzaoui).

rare cases of EC osteomyelitis have been reported in adults in the literature (sternum, base of the skull, clavicle) but to our knowledge, this is the first report of adult EC ischiatic osteomyelitis.

Case report

A 46-year-old patient presented with persistent fever. The patient reported having abdominal pain for 10 days. The clinical examination showed fever 40°C, hepatomegaly 18 cm and splenomegaly 16 cm. Biological test results suggested an inflammatory syndrome: the sedimentation rate in the first hour was 120, CRP: 120 mg/l, polyclonal hypergammaglobulinemia: 27 g/l, normochrome normocytic anemia and hyperleucocytosis: 25 000/mm³ (including 80% polynuclear neutrophils). Doppler ultrasound and CT scan of the abdomen identified "superior mesenteric vein and splenomesenteric branch thrombosis" (Fig. 1).

Other tests to identify the etiology included: infectious serology (atypical germs, Vidal-Wright test, tuberculosis: negative), cytobacteriological urine test (negative), C and S proteins, anti-thrombin II, Leiden Factor V (normal). Blood culture was positive for negative Gram bacillus, without specifically identifying the germ. Dosing for APL showed positive anticardiolipins (Ig G = 19 U/L, Ig M > 100 U/L) positive anti-β 2 GPI antibodies 13 UI/L.

The patient was given two empiric antibiotics associating Amoxicillin + clavulanic acid (Augmentin®) at a dose of 3 g par day, associated with Metronidazole (Flagyl®) at a dose of 1.5 g per day. Fifteen days later while in hospital, the patient developed acute pain in the left hip. Ultrasound of the soft tissues was normal. Pelvic CT scan and NMRI showed a 7 × 4 cm collection along the main axis of the left ischiatic tuberosity extending to the quadratus femoris muscle and the external obturator muscle (Figs. 2–4).

A diagnosis of stage III ischiatic osteomyelitis, complicated by an abscess was made. The patient underwent surgery lying on her back in the lithotomy position, with the legs flexed and in abduction in stirrups and the buttocks on the edge of the table. The approach was direct with an arched incision at the fold of the buttocks at the

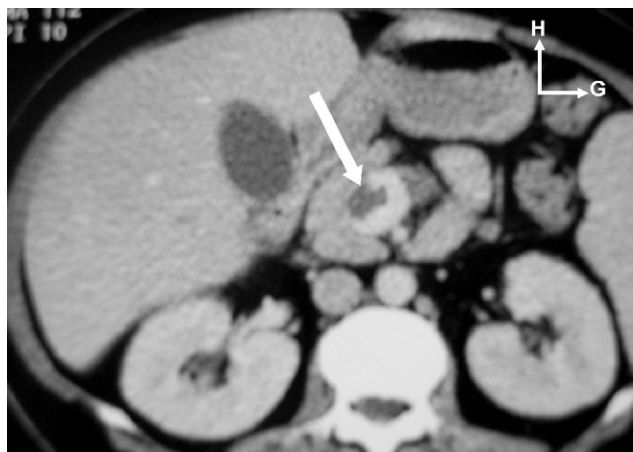


Figure 1 Axial abdominal CT scan after intravenous contrast medium injection: splenomesenteric branch thrombosis.



Figure 2 Axial CT scan of the pelvis with a bone window setting. The cortex of the left ischiatic tuberosity is irregular.

level of the ischiatic tuberosity. A 1 × 3 × 6 cm purulent area was identified on the ischium. A bacteriological sample was taken. The ischium was deperiosteal and irregular. The bacteriological culture confirmed the presence of EC. Antibiotic treatment associated teicoplanin (Targocid®: 400 mg × 2 per day) and imipenem (Tienam®: 1 g × 3 per day). The EC that was identified was resistant to Augmentin®. The clinical and biological picture improved rapidly (normalisation of CRP postoperative D9). Intravenous antibiotics were continued for 21 days, then antibiotics were taken orally: Rifampicin (Rifadine®: 600 mg per day) and Ciprofloxacin (Ciflox®: 1,5 g per day) for two months. The postoperative course was uneventful. A colonoscopy was performed to identify the point of entry and polyps of the colon were identified on anatomopathology as "tubulous adenomas with slight to moderate dysplasia". The APL controls one year later were normal. The abdominal Doppler-ultrasound did not identify any thrombosis.



Figure 3 Axial CT scan of the pelvis after injection of iodated contrast material. Low intensity image of the left external obturator muscle (star) and left quadratus femoris muscle (dot) with ring-like contrast enhancement.

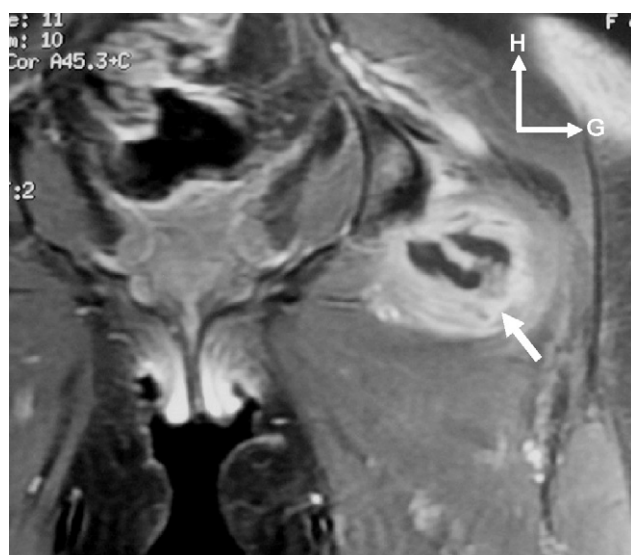


Figure 4 Coronal T1-weighted MRI image with contrast medium. Ring-like enhancement of the left adductor compartment (arrow).

Discussion

Osteomyelites of the pubis and ischium were first described in 1953 by Ingelrans and Lacheretz [1]. In most cases, the lesions begin near the ischiopubic junction [1]. They are generally diagnosed in the late stage with complications: abscess, septicemia, secondary localisations [2].

Ischiatic osteomyelitis, which is usually found in children and adolescents [3], is extremely rare in adults [1]. In general, the local infectious syndrome appears later [2] and the germ is usually *Staphylococcus aureus* (90% of cases), while other entities are less frequent such as *Streptococcus*, *Hemophilus influenzae* or *Salmonellas* [4]. A resistant EC strain may also be found, as in our case, explaining the failure of antibiotic therapy and the development of complications. In our patient, the blood culture identified a negative Gram bacteria, which could not be identified, but it was not anaerobic or *Hemophilus*.

EC osteomyelites are frequently seen in children and the point of entry is usually urinary, but it may also be gastrointestinal. A few rare cases of adult EC osteomyelitis have been reported in the literature (sternum, base of the skull, clavicle) but to our knowledge, EC ischiatic osteomyelitis has never been reported in an adult. The favouring factors may be: immunosuppression, young age or recent surgery (gastrointestinal, urinary) [5]. Identifying the entry point is essential. NMRI is the technique of choice in the diagnosis of pelvic osteomyelitis, in particular ischiatic. [6].

The second specificity of our case was the deep vein thrombosis associated with an APL syndrome. The association of APL antibodies and infection is well known. The pathogen, whether it is viral, bacterial, parasitic or mycotic, plays a role in the development of these auto-antibodies, whose thrombotic effect is still under debate, but which has often been described. EC infection has been found to be the cause of thrombotic accidents in a few cases. Within this framework, Graffin et al. [7] reported a catastrophic case of APL antibody syndrome secondary to EC septicemia from a urinary infection. Other authors explain this association by the fact that EC causes stimulation in domain I of the β 2 glycoprotein I [8]. Thus, our case is original because of the ischiatic location of the osteomyelitis, because of the EC germ, the antiphospholipid antibody syndrome associated with multiple thromboses and because it was secondary septicemia with a gastrointestinal port of entry from colon polyps.

Conclusion

This case reports a rare location of EC ischiatic osteomyelitis in an adult. The difficulties of diagnosis emphasize the importance of performing early MRI. Treatment is based on early antibiotic therapy associated with surgical draining.

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